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Cervical vertebral anomalies in skeletal malocclusions: A cross-sectional study on orthodontic patients at the Aga Khan University Hospital, Pakistan

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Abstract

Introduction: Morphological deviations of the cervical vertebral column have been described in relation to craniofacial aberrations and syndromes. Furthermore, it has recently been shown that abnormal morphology of upper cervical vertebrae is associated with malformation of the jaws and occlusion. Accordingly, it is relevant to focus on similar associations in patients with skeletal malocclusions. Therefore, the objectives of this study are to:

a. Identify the anomalies of the cervical column in patients with skeletal malocclusions
b. Determine the association between cervical vertebral anomalies and skeletal malocclusions.

Materials and Methods: This cross-sectional study was conducted on a total of 90 subjects at the Aga Khan University Hospital, Pakistan. The inclusion criteria were: (1) Pakistani origin; (2) standardized pretreatment profile radiograph with first six cervical vertebrae visible; and (3) accessibility of the second-profile radiograph (mid- or posttreatment). The exclusion criteria were: (1) A prior history of orthodontic treatment; (2) any craniofacial anomaly; and (3) systemic muscle or joint disorder. Lateral cephalograms of all subjects ($n = 90$) were traced by the principal investigator and sagittal jaw relationship was assessed. A total of 30 subjects each with skeletal Class I, Class II, and Class III malocclusions were selected and the cervical vertebral anomalies were observed on their cephalometric radiographs. The frequencies of cervical vertebral anomalies according to skeletal malocclusion categories and gender were analyzed with the Chi-square test, whereas association of cervical vertebral anomalies with skeletal malocclusions was assessed with logistic regression analysis. The level of significance ($P \leq 0.05$) was used for the statistical tests.
Results: Most common anomaly observed in the three groups was fusion between C2 and C3 ($P = 0.006$). This anomaly was found in 20% of subjects with skeletal Class I, 50% of subjects with skeletal Class II and 53.3% with skeletal Class III malocclusions. The highest frequencies of partial cleft at the level of C1 and occipitalization were observed in subjects with skeletal Class II and III malocclusions, respectively. However, none of the subjects showed fusion between C1 and C2 or dehiscence. No statistically significant gender difference was found in the occurrence of morphological deviations of the cervical column. The association of cervical vertebral anomaly was found to be the highest with skeletal Class III and lowest with skeletal Class I malocclusions.
Conclusion: Fusion between C2 and C3 seems to be the most commonly occurring anomaly. This anomaly seems to be more often associated with skeletal Class III than skeletal Class I or Class II malocclusions.

Keywords: Cervical vertebral anomaly, fusion, occipitalization, skeletal malocclusion

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Congenital anomalies of the cervical spine are rare and range in severity from those that are benign or asymptomatic to anomalies that can potentially cause severe complications. Anomalies of the occipito-cervical junction often are not detected until late childhood or adolescence, and some remain hidden well into adult life. Other anomalies of the cervical spine, although recognized in early life, may not become clinically significant until adulthood. Previously, anomalies of the cervical vertebral column were related only with the cleft lip and palate, [1],[2],[3],[4],[5],[6] and craniofacial syndromes. For instance, aberrations of the cervical column morphology were observed in Klippel-Feil, Turner, Saethre-Chotzen, and Down syndromes. [7],[8],[9],[10],[11] Moreover, earlier studies indicated deviations such as fusion anomalies in craniosynostosis syndromes. [12],[13],[14],[15],[16]

Current literature suggests that variations of the cervical column morphology occur in healthy subjects with neutral occlusion and normal craniofacial morphology as well as in orthodontic patients with deviating craniofacial morphology and severe malocclusion. A recent study found that the fusion between the upper cervical vertebrae (C2 and C3) occurred in 14.3% of healthy subjects. [17] Fusions of the upper cervical column within that range are thus considered normal. A great deal of effort has been made to identify the association of cervical vertebral anomalies with abnormal craniofacial morphology, and it was found that the deviation of cervical vertebral morphology is associated with skeletal deep bite, [18] skeletal open bite, [19] skeletal maxillary overjet, [20] skeletal mandibular overjet, [21] , as well as condylar hypoplasia. [17] Deviations occurred significantly more often in all five patient groups compared with the control group.

The precise origin of cervical vertebral anomalies is still unstated, but it has been suggested that the association between abnormal development of cervical vertebrae and the maxilla and the mandible might be caused by a developmental fault of the mesenchyme as these structures might be dependent on the similar para-axial mesoderms. [12],[15],[17] Hence, it is appropriate to focus on this area of research and to consider the craniofacial morphology as an important diagnostic tool in orthodontic treatment planning. To the best of our knowledge, no local data are available in this regard.

Our aims in this study were therefore (1) to identify the anomalies of the cervical column in Pakistani patients with skeletal malocclusions and (2) to determine the association between cervical vertebral anomalies and skeletal malocclusions.

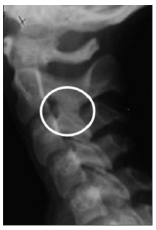
MaterialS and Methods

The sample of this cross-sectional study was selected from the records of patients seeking orthodontic treatment (n = 90) at the Aga Khan University Hospital, Pakistan. The inclusion criteria were: (1) No prior history of orthodontic treatment; (2) no craniofacial anomaly; (3) no systemic muscle or joint disorder; (4) standardized pretreatment profile radiograph with first six cervical vertebrae visible; and (5) accessibility of the second-profile radiograph (mid- or posttreatment).

Lateral profile radiographs were taken by the method described by Siersbaek-Nielsen and Solow, [22] that is, teeth in occlusion, standardized head posture and the mirror position. The radiographs were taken in a cephalostat (Orthoralix 9200/Ceph) with a film-to-focus distance of 134 cm and a film-to-median plane distance of 15 cm. The pretreatment lateral cephalograms were selected and traced by the principal investigator. The sagittal jaw relationship was assessed by measuring the ANB angle and evaluating the McNamara and Wits analyses. The total sample (n = 90) including 51 females (mean age 15.6 years, SD ± 6.00) and 39 males (mean age 15.3 years, SD ± 4.95) was categorized into skeletal Class I, Class II, and Class III categories according to the sagittal jaw relationship. The Class I malocclusion group comprised of 30 subjects (17 females and 13 males) with a sagittal jaw relationship ranging 1-4° and dentoalveolar horizontal maxillary overjet of 1-5 mm, the Class II malocclusion group included 30 subjects (20 females and 10 males) with a sagittal jaw relationship > 5° and dentoalveolar horizontal maxillary overjet of 6-12 mm, whereas the Class III malocclusion group comprised of 30 subjects (14 females and 16 males) with sagittal jaw relationship of < 0° and dentoalveolar horizontal mandibular overjet of 0-5 mm. To assess the morphology of the cervical column, the first six cervical vertebrae were visually inspected on standardized lateral skull radiograph film as well as confirmed digitally using Rogan View Pro-X (version 4.0.6.2, PACS/RIS/XDS solutions by Rogan-Delft, Netherlands). Characteristics of the cervical column were classified into two broad categories:

- Fusion anomalies, labeled as fusion, block fusion, and occipitalization. Fusion is defined as a merger of one unit with another at the vertebral bodies, articulation facets, neural arch, or transverse processes [Figure 1]. Occipitalization is defined as a union, either partially or completely, of the atlas (C1) with the occipital bone [Figure 2]. Block fusion is defined as a fusion of more than two units at the vertebral bodies, articulation facets, neural arch, or transverse processes

Figure 1: Illustration of fusion of the C2 and C3 vertebrae



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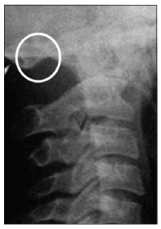

Figure 2: Illustration of occipitalization between the C1 vertebra and the occipital bone

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- Posterior arch deficiency is subdivided into partial cleft and dehiscence. Partial cleft is defined as failure of the posterior part of the neural arch to fuse [Figure 3], and dehiscence is defined as failure of a part of a vertebral unit to develop.

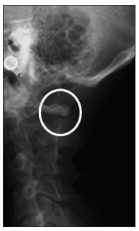

Figure 3: Illustration of partial cleft of posterior part of neural arch of the atlas

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The mid- or posttreatment profile radiographs were used to confirm the observations and only anomalies verified on these second radiographs were registered as anomalies of the cervical vertebral column.

Statistical analysis

The intra-examiner reliability was assessed by randomly selecting 10 radiographs from the study sample and visually re-evaluating the morphology of cervical vertebrae. However the Pearson's correlation showed a strong correlation between the two sets of observations (r = 0.82, P = 0.03). The frequency of cervical vertebral anomalies in the three groups and in both genders was assessed with the Chi-square test. A logistic regression analysis was performed to determine the association of cervical vertebral anomalies with skeletal malocclusions. The statistical analyses were performed with SPSS (version 19.0, SPSS Inc., Chicago, IL, USA). The level of significance ($P \leq 0.05$) was used for the statistical tests.

Results

A statistically significant proportion of the total sample showed fusion between C2 and C3 (P = 0.006). Partial cleft at the level of C1 was the second common anomaly (P = 0.090) followed by occipitalization (P = 0.160). None of the subjects showed fusion between C1 and C2 or dehiscence. The detailed frequencies of observed anomalies are presented in [Table 1]. No statistical gender differences were observed in the distribution of different anomalies. The frequency of fusion between C2 and C3 is shown in [Table 2]. The odds of this anomaly are 4 times higher in Class II category than in Class I category, and 4.5 times higher in Class III category as compared to Class I category.

Table 1: Frequencies of cervical vertebral anomalies according to skeletal malocclusions categories and gender

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Table 2: Association of fusion between C2 and C3 vertebrae with skeletal Class I, Class II, and Class III malocclusion categories

Skeletal malocclusion (n=90)	Fusion between C2 and C3 (count (%))		*P* value	Exp (B)
	No	Yes		
Class I (n=30)	24 (80.0)	6 (20.0)	0.021	1.000
Class II (n=30)	15 (50.0)	15 (50.0)	0.018	4.000
Class III (n=30)	14 (46.7)	16 (53.3)	0.009*	4.571

n=90. Chi-square test. Logistic regression analysis. *P≤0.05, *P≤0.001=Highly statistically significant. Exp (B)=Exponentiation of B coefficient (OR). OR=Odds ratio

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Discussion

Our study aimed to identify the commonly occurring anomalies as well as the association of these anomalies with

skeletal malocclusions. The results suggest that the most frequently occurring anomaly was fusion between C2 and C3 (P = 0.006). A descending order of association of this anomaly was established with skeletal Class III, Class II, and Class I malocclusions, respectively. The pattern of cervical vertebral deviation was in concordance with the previous studies of orthodontic patients with severe maxillary [20] and mandibular overjet. [21] These studies showed that the fusion occurred in 14-21% of control groups, 52% of patients with severe maxillary overjet and 61% of patients who had severe mandibular overjet. In all these study groups, fusions were always observed between the second and third cervical vertebrae.

In addition, partial cleft at the level of C1 and occipitalization occurred with a higher frequency compared to the rest of the cervical column deviations. However, dehiscence and fusion between C1 and C2 were not observed among the subjects at all. Previous studies [18],[19],[20],[21] did not report partial clefts and occipitalization as common findings and hence contrast with the results of the present study.

Although not all results were significant, the overall direction showed that deviations of cervical vertebral column occurred significantly more often in skeletal Class II and Class III groups than in Class I group. This was expected because previous studies showed a higher prevalence of cervical vertebral anomalies in subjects with severe skeletal malocclusion. [18],[19],[20],[21] The occurrence of these deviations in the cervical column and their different prevalence in skeletal malocclusion groups are still inexplicable. An explanation for this could be the involvement of notochord in the development of cervical vertebral bodies and basilar part of occipital bone (posterior part of cranial base angle) during the early prenatal period. [23],[24],[25],[26],[27],[28],[29] Since the cervical vertebrae and cranial base have similar embryonic origin, and the jaws are attached to the cranial base, the cranial base could be considered as a developing link between the cervical vertebral column and the jaws. [30] Hence, fusion anomalies as seen in our subjects could be signs of deviations in the early development or signaling between the notochord, neural tube, neural crest cells, and para-axial mesoderm. Therefore, the clinical significance of such an association is early diagnosis and correct treatment of these patients.

Sonnesen et al. [17] had reported that the deviations in head posture and cranial base angle were sexually dimorphic, showing larger cervicohorizontal and cranial base angles in females than males. They also observed a positive correlation of cervical lordosis, inclination of upper cervical spine and cranial base angle in females with fusions of the cervical column, whereas this correlation was not found in males. Hence, it could be hypothesized that fusion anomalies show dimorphic pattern in their occurrence. However, Sonnesen and Kjaer [18] and Arntsen and Sonnesen [31] proposed that there was no significant gender difference in the occurrence of cervical vertebral anomalies. The present study supports this finding showing no gender dimorphism.

One limitation in our study was the use of two-dimensional lateral radiographs to record the deviations of cervical vertebral anomalies. This was performed to avoid any additional radiographic exposure to the subjects, as lateral profile radiograph is a routine investigation required for orthodontic treatment planning. However, second-profile radiographs were used to rule out any observational error, and the findings were reconfirmed by using a digital radiographic viewer.

Conclusions

- Cervical vertebral anomalies occur significantly more in subjects with skeletal Class II and Class III malocclusions as compared to skeletal Class I malocclusion
- No gender dimorphism is observed in the occurrence of cervical column deviation
- Most frequently occurring anomaly is fusion between C2 and C3
- Association of fusion between C2 and C3 is highest with skeletal Class III malocclusion and lowest with skeletal Class I malocclusion
- Congenital anomalies of the cervical column are worthy of attention.

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Figures

[Figure 1], [Figure 2], [Figure 3]

Tables

[Table 1], [Table 2]

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